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CASE REPORT

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# Rupture of hidden abnormal myometrial vessels during cesarean delivery of a patient with subserosal leiomyoma: A possible pathogenesis of sudden-onset disseminated intravascular coagulation

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**Key Clinical Message**

We report a case of sudden-onset disseminated intravascular coagulation during cesarean delivery for a patient with a subserosal leiomyoma. Rupture of hidden anastomotic vessels resulted in a significant decrease in fibrinogen levels and uncontrolled bleeding. Uterine venous flow disturbance caused by subserosal leiomyoma compression can possibly cause such a situation.

**KEYWORDS**

disseminated intravascular coagulation, fibrinogen, hysterectomy, leiomyoma, obstetric hemorrhage

## 1 | INTRODUCTION

Uterine atony, blood vessel injury, and coagulation disorders are the major causes of life-threatening postpartum hemorrhage. Depletion of coagulation factors caused by an underlying condition such as amniotic embolism prior to or during labor may worsen the situation and contribute to overt obstetric disseminated intravascular coagulation (DIC) and uncontrollable obstetric hemorrhage. In such cases, prompt transfusions, administration of coagulation supplements, and prompt surgical treatment are often required. However, antenatal examinations to predict such severe complications remain difficult. We report a case of sudden-onset DIC caused by the rupture of hidden abnormal myometrial vessels during cesarean delivery for a patient with a subserosal leiomyoma.

## 2 | CASE EXAMINATION

A 39-year-old primiparous woman was admitted for an elective cesarean delivery due to the obstruction of the birth canal caused by a subserosal leiomyoma in the lower uterine segment. She had a history of Hashimoto disease and idiopathic thrombocytopenia purpura (ITP). During the pregnancy, routine ultrasound examinations did not reveal any uterine abnormalities except the leiomyoma; color Doppler ultrasonography to screen for abnormal myometrial vessels was not considered required. Her thyroid disorder was well controlled with oral levothyroxine. Her platelet count was within 28-158 $\times 10^9$/L, but it increased to 192 $\times 10^9$/L after 5 days of γ-globulin (400 mg/kg/d) administration prior to the surgery. Her hemoglobin level was 9.6 g/dL. Therefore, at 37 weeks and 5 days of gestation, cesarean delivery was

performed under spinal anesthesia. During surgery, the anterior surface of the uterine wall was visually normal. However, vigorous venous bleeding from the incision site was observed immediately. A healthy male newborn with Apgar scores of 7 and 8 at 1 and 5 minutes, respectively, weighing 2452 g was delivered. The incised uterine wall was immediately closed in two layers after the placenta was removed. Intravenous oxytocin and methylergometrine were administered to prevent postpartum hemorrhage; however, the incision site expanded irregularly and a hematoma within the anterior wall of the uterine body became obvious (Figure 1). Despite additional sutures placed in the anterior uterine wall, the size of the expanding hematoma continued to increase. The estimated blood loss increased to 1530 mL, and her blood pressure suddenly decreased to 70/30 mm Hg. To prevent uncontrolled bleeding, an emergency supracervical hysterectomy procedure was performed. Subsequently, 12 units of packed red blood cells, 10 units of fresh-frozen plasma, 30 units of platelets, and 4 g of fibrinogen concentrate were administered for hemostasis. The total estimated blood loss was more than 4 L by the end of the surgery. Laboratory examination prior to the hysterectomy revealed that the patient's hemoglobin level had decreased to 7.0 g/dL, platelet count decreased to $51 \times 10^9$/L, prothrombin time was 16.2 seconds, and fibrinogen level had significantly decreased to 59 mg/dL.

Grossly, the uterine specimen included the uterus without the cervix and the subserosal leiomyoma on the left lower trunk of the uterus. Numerous abnormally dilated vascular lacunas (Figure 2) were identified within the anterior myometrial wall across the incision site (Figure 3A). Postoperative histological examination revealed lymph ducts and markedly dilated veins consisting of a single endothelial layer (Figure 3B). However, these findings were not consistent with the characteristics of arteriovenous malformation, varices, or cavernous hemangiomas. Therefore, a dilated anastomotic vascular network was the most probable cause. Postoperatively, ventilator support was administered to the patient in the intensive care unit until postoperative day 2. Her anemia and coagulation disorder improved after massive transfusions, and she was discharged on day 7.

## 3 | DISCUSSION

### 3.1 | How did the abnormal vessels arise?

This case of sudden-onset DIC was caused by the rupture of hidden abnormal myometrial vessels during cesarean delivery for a patient with subserosal leiomyoma. Emergency supracervical hysterectomy and massive transfusion were required. Postoperative pathological examination excluded the possibility of arteriovenous malformation, varices, and cavernous hemangiomas. Therefore, we believe that the abnormal vessels in the anterior myometrial wall were part of the dilated anastomotic vascular network, and they compensated for the left uterine venous flow disturbance caused by subserosal leiomyoma suppression during the pregnancy.

### 3.2 | How can it be predicted?

Blood flow in the uterus increases during pregnancy. Furthermore, the leiomyoma increases in size and expands its feeding vessels. Although postpartum hemorrhage due to uterine atony and dysfunctional labor are commonly reported[1] and are associated with large leiomyomas,[2] most are considered intramural myomas and not subserosal leiomyomas, as in this case. Although this is a single-case experience, we assume that the DIC in

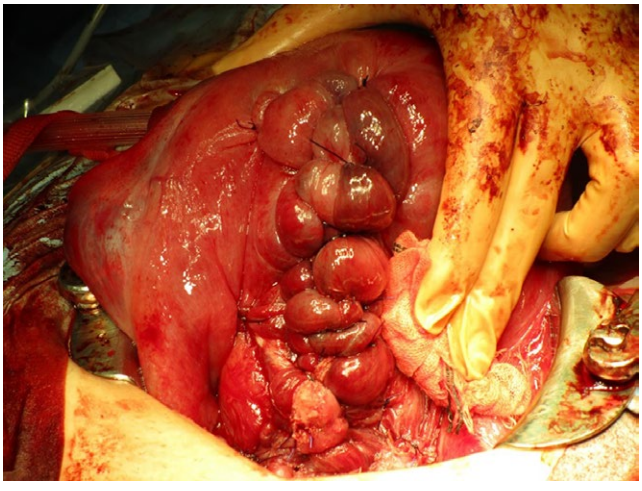

**Figure 1.** The incision site expanded irregularly after the suturing of the anterior uterine wall to control the expansion of the hematoma

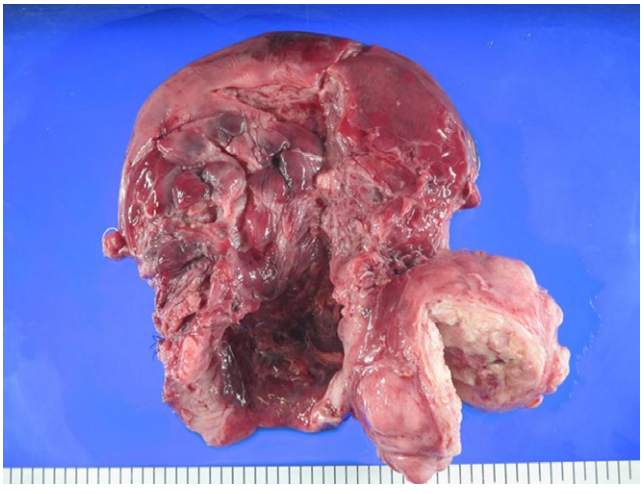

**Figure 2.** The specimen included the supracervical uterus and subserosal leiomyoma. Numerous lacunae were identified within the myometrial wall

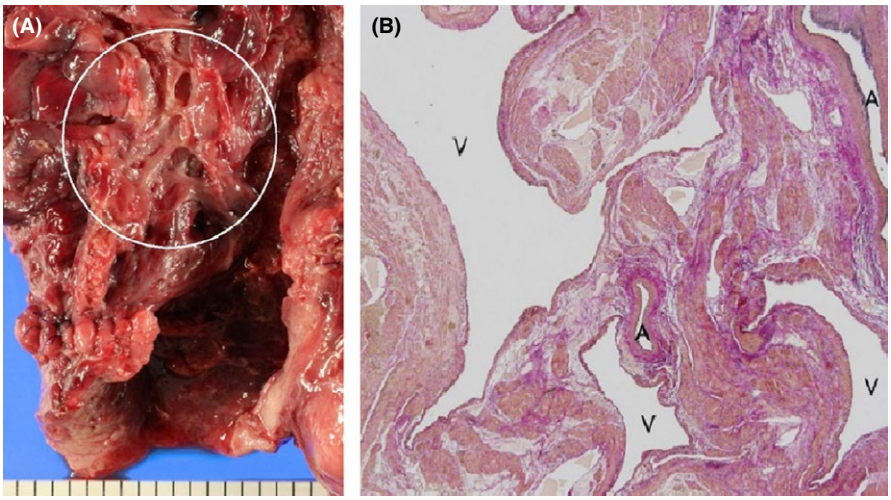


**Figure 3.** A, Postoperative pathological examination revealed irregularly dilated vessels within the uterus, at the incision site. B, Numerous dilated vein structures consisting of a single layer of epithelial cells were identified (Elastica-van Gieson staining, ×100). A, artery; V, vein

the present case was mainly caused by the rupture of hidden vessels during the cesarean delivery. Therefore, antenatal Doppler ultrasonography of the anterior uterine wall might be useful for predicting DIC in cases where vascular abnormalities are suspected, before cesarean delivery.

### 3.3 | How should it be treated?

Most abnormal myometrial vessels are difficult to treat using partial sectioning and suturing during cesarean delivery because most of these vessels lack sufficient connective tissue.[3] Moreover, myometrial contractions that are necessary to control uterine blood flow were insufficient in most cases. However, during the cesarean delivery in the present case, fibrinogen level suddenly decreased to 59 mg/dL, which was much lower than that needed to diagnose overt DIC (≤150 mg/dL).[4] We believe this was because the ruptured dilated veins due to the myometrial sectioning were exposed to amniotic fluid, resulting in a DIC type of amniotic fluid embolism,[5,6] although there were no severe cardiopulmonary symptoms involved in this case. Previous studies indicated that fibrinogen levels <87 mg/dL were associated with a very high rate (95.2%) of DIC requiring massive transfusions.[7] Therefore, the extremely low fibrinogen levels found in similar cases might be a key signal indicating the need for prompt transfusion and surgical treatment. Uterine artery embolization might be an option for treating obstetric hemorrhage. However, vasospasm, hemodynamic shock, and DIC may cause failed embolization.[8-10] It is unclear whether the present case would be a good candidate for uterine artery embolization because the subserosal leiomyoma was very close to the uterine artery. However, certain transfusions to correct coagulation disorders prior to the procedure are required.

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## CONFLICT OF INTEREST

None declared.

## AUTHORSHIP

LW, HK, TK, and KT: involved in writing the manuscript. JU, IH, SS, and KC: involved in the management of the patient. AT: involved in the pathological diagnosis.

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